

Cystic retroperitoneal inflammatory myofibroblastic tumor: A case report and review of literature

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ABSTRACT: Inflammatory myofibroblastic tumor (IMT) also known as inflammatory pseudo tumor is an uncommon tumor with controversial etiology and unpredictable biological behavior. They can be found at any anatomic site with a predilection for the lung, the genito-urinary tract and the mesentery. Retroperitoneal location has been rarely reported. This tumor is typically circumscribed, non-encapsulated and often multinodular with solid and firm cut surface. **Presentation:** we report the case of a 56 years old woman with history of abdominal pain since 2 months. Compound tomography (CT) demonstrated a cystic mass on retroperitoneum. The patient underwent surgical resection and a multicystic gray white mass which was well localized in the retro peritoneum was observed. **Discussion:** A review of the literature on IMT yielded only a few number of cystic presentation.

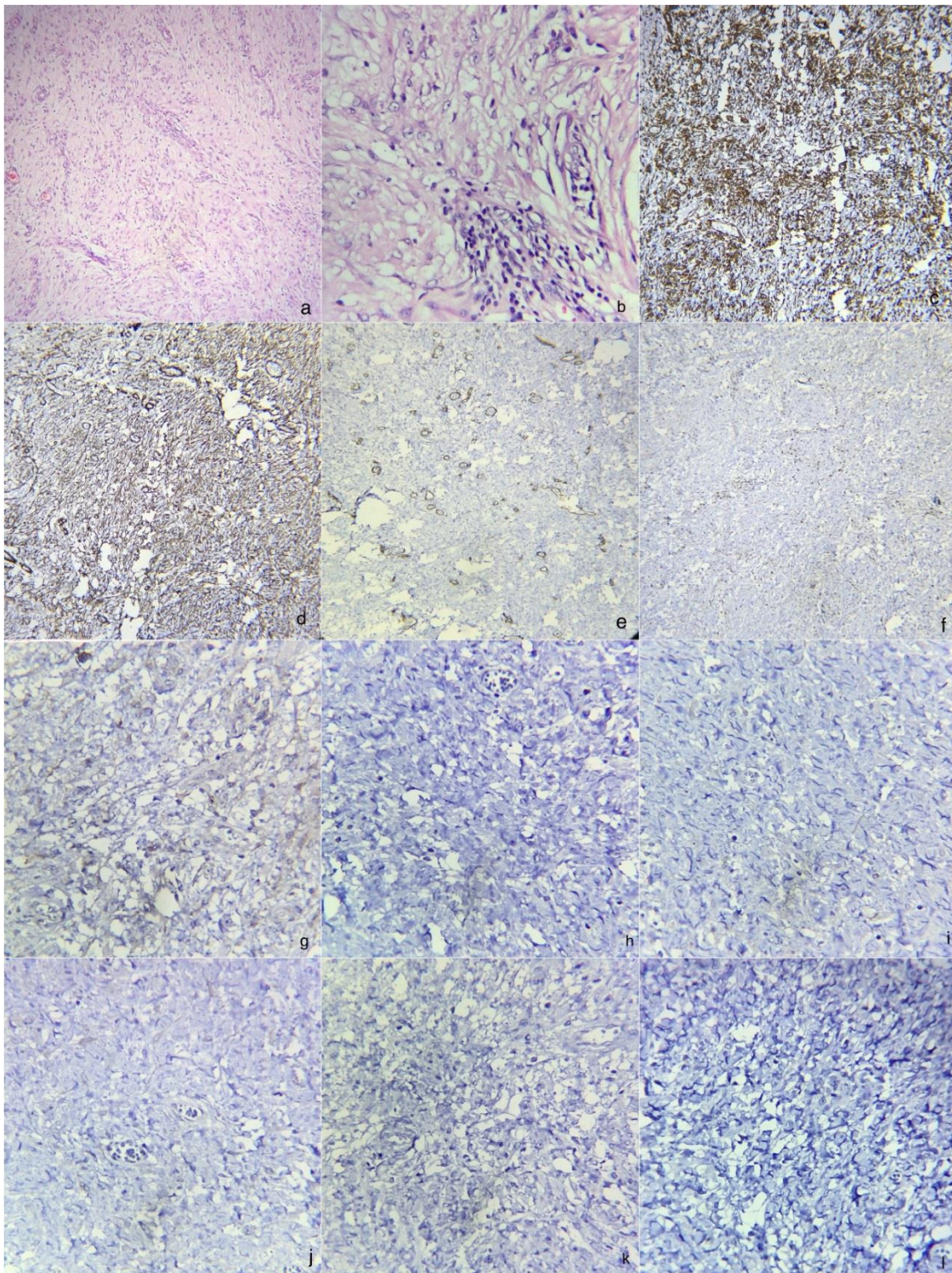
Keywords: Retro peritoneum, Inflammatory, Myofibroblastic, Tumor.

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) were originally termed by Bunn in 1939[1] and is now the generally accepted term for the majority of lesions formerly named inflammatory pseudo tumor, plasma cell granuloma, mesenteric myxoid hematoma and inflammatory fibrosarcoma. This unification was achieved on the basis of considerable morphologic and clinical overlap combined with both clinical and genetic evidence of their neoplastic nature[2]. IMT may involve any organ but commonly occurs in the lung or orbit, the most common sites of extra pulmonary inflammatory myofibroblastic tumors are the mesentery and omentum [3,4]. The most accepted theory of pathogenesis supports immunological factors and a post-inflammatory reparative process due to surgery, infection or trauma. The clinical presentation differs depending on the anatomical regions examined. It is often associated with fever, weight loss and anemia [5]. In general, masses appear on gross examination as a nodular and solid sometimes with features of necrosis and hemorrhage [6]. The tumors are essentially cellular, fascicular, fibroblastic/myofibroblastic proliferations accompanied by a prominent infiltrate of chronic inflammatory cells, particularly plasma cells. The immunohistochemistry as with other myofibroblastic lesions are generally actin positive and may also show staining for desmin and keratin [8]. Approximately 50% of inflammatory myofibroblastic tumors harbor clonal rearrangement of ALK gene at 2p23[9,10]. The WHO classification places inflammatory myofibroblastic tumors in an intermediate category because of tendency for a small risk of metastases and local recurrence [11,12,13]. These findings have recently shown that chromosomal abnormalities may be suggestive of clonal origin, not merely a reactive process and should be considered as a true neoplasm[12,13]

Case presentation: we report a 56 year old female presented with 3 month history of abdominal pain. Computed tomography (CT) demonstrated cystic mass on retro peritoneum. The macroscopic feature was multicystic cream-brown mass measuring 16×12×5cm with wall thickness about 1.8cm. The histological study revealed a spindle tumor with multicystic spaces without any lining epithelium which show fibroblastic/myofibroblastic infiltration in vascularized, myxoid, edematous and inflammatory background. Immunohistochemistry showed that the tumor cells were positive for

smooth muscle actin (SMA), Desmin and some tumor cells for ALK, but negative with S-100,CD34,CK,EMA,CD117[figure] . These results confirmed the diagnosis of IMT.



A)inflammatory my fibroblastic tumor(IMT). B)my fibroblasts in inflammatory background. C)smooth muscle actin(SMA).
D)Desmin. E)CD34. F)Bcl2. G)ALK. H)CK. I)Ki67. J)EMA. K)S100. L)Cd117

Discussion: inflammatory myofibroblastic tumors (IMT) is a rare neoplasm of intermediate malignant potential characterized by myofibroblastic proliferation and mixed inflammatory cell infiltrate. The etiology of inflammatory myofibroblastic tumors is unknown while some authors propose a neoplastic origin, others believe that it is an immunological response to an infectious or inflammatory process. The tumor commonly occurs in lung and extra-pulmonary IMTs are rare. These tumors form whorled firm white or yellow-colored fleshy mass. Secondary changes include hemorrhage, necrosis, calcification and ossification. In the largest series of 84 cases of extra-pulmonary IMT, only four retro-perineal IMT were reported and all of 84 cases were firm and solid with infiltrative borders [14]. Ntloko and Gounden reported 5 patients with intestinal IMT, one tumor had soft myxoid consistency with cystic degeneration and four tumors had solid firm appearance on cut section [15]. Shilou Feng and Aixiang Wang reported inflammatory myofibroblastic tumor as renal cyst [16]. In our case myofibroblastic tumor appears with cystic mass which is uncommon feature.

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